

Incidence of Aneurysm Formation After Dacron Patch Aortoplasty Repair for Coarctation of the Aorta: Long-Term Results and Assessment Utilizing Magnetic Resonance Angiography With Three-Dimensional Surface Rendering

WILLIE JAMES PARKS, MD, THANG D. NGO, BA, WILLIAM H. PLAUTH, JR., MD, FACC,*
ESTELLE R. BANK, MD,† SCOTT K. SHEPPARD, MS,‡ RODERIC I. PETTIGREW, PhD, MD,†
WILLIS H. WILLIAMS, MD†

Atlanta, Georgia

Objectives. Magnetic resonance angiography with three-dimensional surface rendering was performed to determine its value in assessing anatomic detail in patients with suspected aortic aneurysms.

Background. Dacron patch aortoplasty repair of coarctation of the aorta carries an inherent risk of aneurysm development. Sudden death from aortic rupture prompted discontinuing this operation and evaluating 39 patients (16 girls; mean age 6.3 years, range 10 days to 14.5 years) undergoing repair between January 1976 and October 1987. The aorta ruptured in 10 patients; 6 died at a mean interval of 8.1 years (range 0.75 to 12.4) after repair. All 33 survivors were interviewed and examined.

Methods. Conventional magnetic resonance imaging was performed in 26 patients, magnetic resonance angiography in 18. Angiographic slices were used to reconstruct three-dimensional images. No catheterization or contrast angiography was per-

formed. Surgical intervention was based on clinical findings and magnetic resonance images.

Results. Twenty patients (11 girls) developed aneurysms, of which nine were detected in patients studied by magnetic resonance. Ruptures occurred in eight female patients, three of whom were pregnant. Surface renderings accurately defined aortic anatomy or aneurysms in all patients. On follow-up, no aneurysms have been detected in patients with negative magnetic resonance study results. Precise anatomic correlation with operative findings was reported.

Conclusions. Magnetic resonance angiography with three-dimensional surface rendering provides noninvasive, radiation-free and contrast agent-free high resolution images of the thoracic aorta. These images can be reviewed and have three-dimensional form and perspective. These techniques were preferred over invasive angiography by surgeons and clinicians as definitive, risk-free procedures before surgical intervention.

(*J Am Coll Cardiol* 1995;26:266-71)

Dacron patch aortoplasty gained popularity in the mid-1960s because of its excellent gradient resolution and because it avoided sacrifice of intercostal arteries and minimized restenosis by eliminating circumferential suture lines (1). During this period, Dacron patch aortoplasty technique was extensively used by many institutions for repair of coarctation of the aorta. Late aneurysm formation, a most serious postoperative and life-threatening complication, has led many institutions, including our own, to abandon the technique (2,3). The incidence of aneurysm formation after Dacron patch aortoplasty

in reported series varied from 4% to 38% (4-10). In this review, we report a 51% late aneurysm formation after Dacron patch aortoplasty repair of coarctation of the aorta and describe the utilization and usefulness of magnetic resonance imaging and three-dimensional magnetic resonance angiography in perioperative evaluation of patients.

Methods

Patients. Thirty-nine patients underwent Dacron patch aortoplasty repair of coarctation of the aorta at Emory University between January 1976 and October 1987. Dacron patch aortoplasty was the initial operative procedure for native coarctation of the aorta or was performed during a required revision by three surgeons at Emory University. Thirty-three patients initially underwent Dacron patch aortoplasty only; six underwent varied primary procedures: end-to-end repair (three patients), a combination of subclavian flap aortoplasty and end-to-end repair (one patient), end-to-end repair and

From The Children's Heart Center: *Egleston Children's Hospital at Emory University; and †Departments of Medical Systems and Radiology, Emory University Hospital, Atlanta, Georgia. This study was supported in part by the Children's Research Center at Emory University, Atlanta, Georgia.

Manuscript received June 7, 1994; revised manuscript received February 3, 1995, accepted February 27, 1995.

Address for correspondence: Dr. Willie James Parks, The Children's Heart Center at Emory University, 2040 Ridgewood Drive, Northeast, Atlanta, Georgia 30322.

Table 1. Results by Age Group at Time of Operation

	Age At Operation	No. (%) of Aneurysms	No. of Deaths	No. of Ruptures
Group I (9 patients)	<2 yr* (0.33/0.48)†	1 (11%)	1	1
Group II (30 patients)	>2 yr (7.9/3.34)†	19 (63%)	5	9
Total	—	20 (51%)	6	10

*Two-sample test, 99% confidence interval ($p < 0.01$). †Mean value/SD.

Dacron patch aortoplasty (one patient), and subclavian flap aortoplasty and Dacron patch aortoplasty (one patient). Five of these six patients subsequently underwent revision of their repair for recoarctation of the aorta. Age at Dacron patch aortoplasty ranged from 10 days to 14.5 years (mean 6.3 years).

Study groups. The patients were classified into two groups on the basis of age at Dacron patch aortoplasty repair of coarctation (Table 1): repair at <2 years of age (group I, $n = 9$) and repair at >2 years of age (group II, $n = 30$). In group I, eight patients underwent Dacron patch aortoplasty between 10 days and 7 months of age; one other patient had repair at 1.5 years of age (mean 0.33 years, SD 0.48). Age at operation in group II ranged from 3.5 to 14.5 years (mean 7.9, SD 3.34). Follow-up ranged from 12.6 to 17 years (mean 15, SD 1.7) and 6 to 16 years (mean 13.3, SD 3.1, excluding one patient who died 1 year postoperatively) for groups I and II, respectively. The difference between the mean age at operation and incidence of aneurysm formation between the two yielded a p value <0.01.

Imaging techniques. Conventional electrocardiographic (ECG)-gated spin-echo magnetic resonance imaging (MRI) was performed in all 26 patients. In addition, ECG-gated inflow magnetic resonance angiography with trigger delay and a user-defined diastolic acquisition gated width, known as "gated sweep" (11), was performed in 18 patients. Gated sweep is a technique used to image flow in vascular structures during diastolic filling to minimize the effect of turbulence on the image quality. Figure 1 (top) illustrates a gated sweep image. A series of sequential transverse images of this type spanning the thoracic aorta are acquired in a sequential format and are used to reconstruct three-dimensional images of the aorta. Conventional MRI was also performed and included static spin echo images (Fig. 1, bottom) and, in selected cases, cine fast gradient echo images in transverse, coronal and sagittal planes. The spin echo images were 5 to 8 mm thick with a 10% gap.

The gated sweep images were 3 to 4 mm thick with a 1-mm overlap. Both gated sweep and conventional MRI were performed utilizing a 1.5-tesla ACS Gyroscan (Philips Medical System). Data acquired from each patient were postprocessed on a Philips Gyroview work station with three-dimensional surface-rendering capability (ISG Allegro) for reconstruction of the aorta. The three-dimensional reconstructed images of the aorta were reviewed from different rotational angles and printed on a Kodak thermal printer (XL 7700) for subsequent

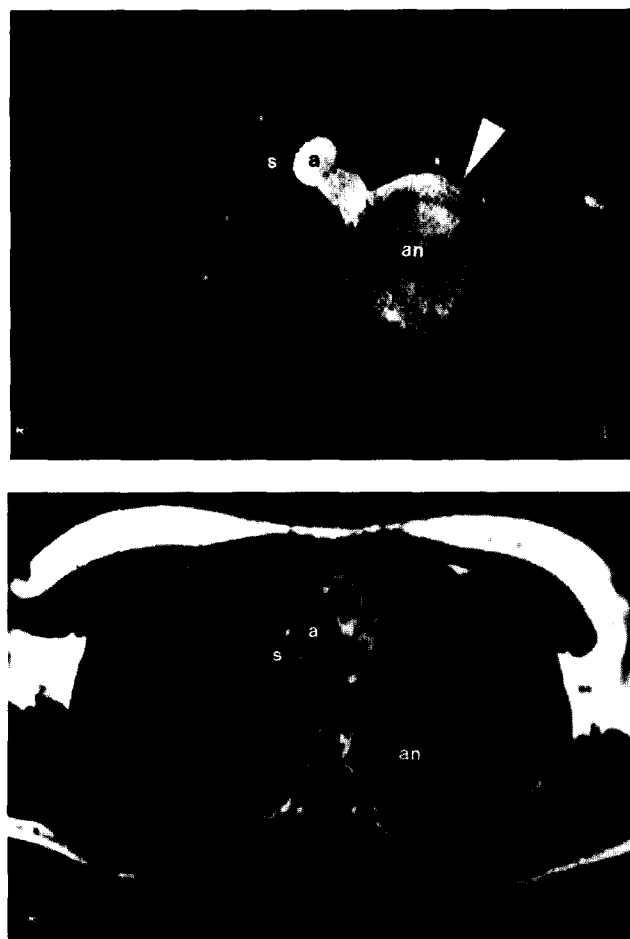


Figure 1. Top, Large aortic aneurysm (an) in a 3-mm transverse slice obtained by the gated sweep technique. The angiographic image outlines the lumen of the vessel at the level of the transverse arch. The ascending aorta (a) and superior vena cava (s) are also shown. Arrow = large saccular aneurysm at the previous Dacron patch aortoplasty repair site. Bottom, Static spin-echo image from the same patient. This transverse image is within several millimeters of the top image. The static spin-echo images do not illustrate flow in the lumen but depict the vascular margins of the aneurysm and other vascular lumens. L = left; R = right.

review and examination. Each study took ~60 to 90 min. Postprocessing time required an additional 20 to 30 min. An aneurysm was diagnosed when the ratio of diameter of the aorta at the repair site to the diameter of the aorta at the level of the diaphragm exceeded 1.5 (3,4,9). Although the definition of an aneurysm is somewhat arbitrary, there is, however, no question about defining an aneurysm in our study patients, as shown by Figure 1.

Statistical methods. All statistical analysis was performed using the Student t test (Origin, MicroCal Software). A p value <0.05 was considered statistically significant.

Results

Twenty aneurysms occurred in 39 patients (51%) (Table 1). Nine of the 20 aneurysms were detected by MRI and magnetic

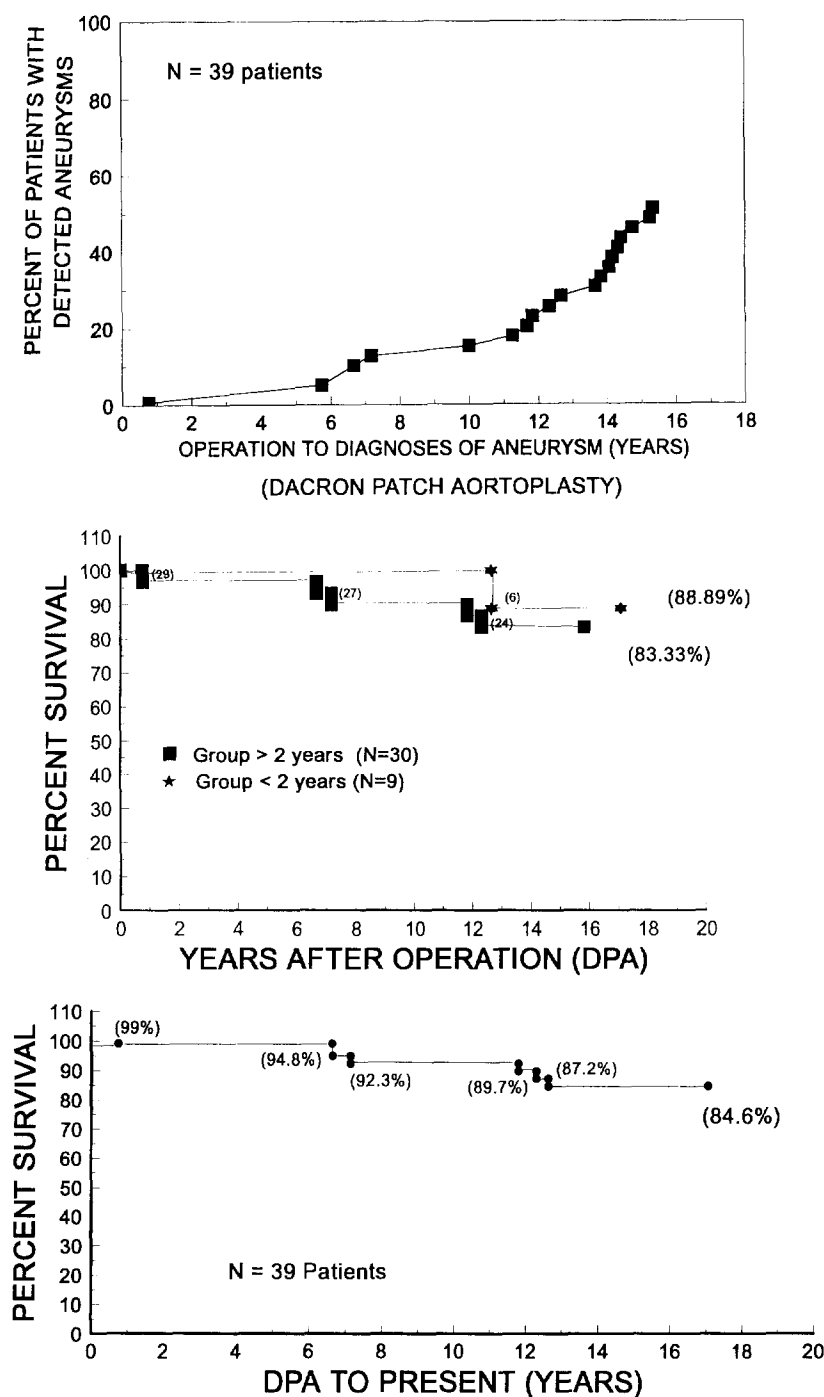


Figure 2. Top, Fifty-one percent of aneurysms were detected >10 years after Dacron patch aortoplasty repair. The frequency of aneurysm formation appears to increase exponentially after 10 years. **Middle,** Group survival rate. Nine patients underwent Dacron patch aortoplasty (DPA) at <2 years of age (group I); one died, yielding a survival rate of 88.9%. The survival rate was 83.3% for patients >2 years old at Dacron patch aortoplasty. **Bottom,** Overall survival rate is 85%.

resonance angiography at our institution; 5 were detected and resected at other institutions, and 6 were diagnosed at autopsy. The rate of detection (years) of aneurysms postrepair are depicted in Figure 2 (top). The increasing frequency of aneurysms appears to progress exponentially after 10 years postoperatively. Ten ruptures occurred in the 20 patients with aneurysms (50%) at the site of previous coarctation repair by Dacron patch graft and were fatal in two male and four female patients (Table 2). Nine (39%) of the 23 male patients had aneurysms versus 11 (70%) of the 16 female patients. Of the 10

patients with aortic aneurysm rupture, 3 were pregnant, and 7 complained of hemoptysis and paraspinal back pain before the event. There were four survivors of aortic rupture; all were female, and two were pregnant. Only one patient (11%) was found to have an aneurysm in group I compared with 19 in group II (63%).

Four female and two male patients died (Table 2) of aortic aneurysm rupture (five in group II, one in group I). One patient died suddenly at her private residence; one died at the beginning of her third trimester of pregnancy; and two others

Table 2. Results of Dacron Patch Angioplasty

Gender	No. of Patients	No. (%) of Aneurysms	No. of Ruptures	No. of Deaths
Male	23	9 (39%)	2	2
Female	16	11 (69%)	8	4*
Total	39	20 (51%)	10	6

*Three pregnancies.

died in the operating room after the aneurysms had ruptured. Autopsy reports of all six patients revealed the cause of death to be aortic rupture (death occurred at a mean interval of 8.1 years [range 0.75 to 12.4] after repair) at the site of the patch aortoplasty. All patient deaths in group II had coarctation of the aorta as the primary diagnosis, whereas the patient death in group I had a ventricular septal defect, hypoplastic arch and coarctation of the aorta. Fourteen patients in group II (>2 years old at operation) had aneurysms that were successfully detected and resected (nine at Emory University Hospital, five elsewhere). Surgical intervention for the nine patients at Emory was planned on the basis of magnetic resonance images and three-dimensional magnetic resonance surface rendering (Fig. 3) alone; no catheterization or contrast angiography was performed.

Surgeons reported precise anatomic correlation between the three-dimensional reconstructed images and the operative findings in each case. The postoperative survival rates (Fig. 2) were 88.89% and 83.33% for groups I and II, respectively, and an overall 84.6% for the combined patient groups.

Symptoms and laboratory findings. Paraspinal back pain and hemoptysis/hematemesis were the most often reported symptoms during historical review of patients with aortic

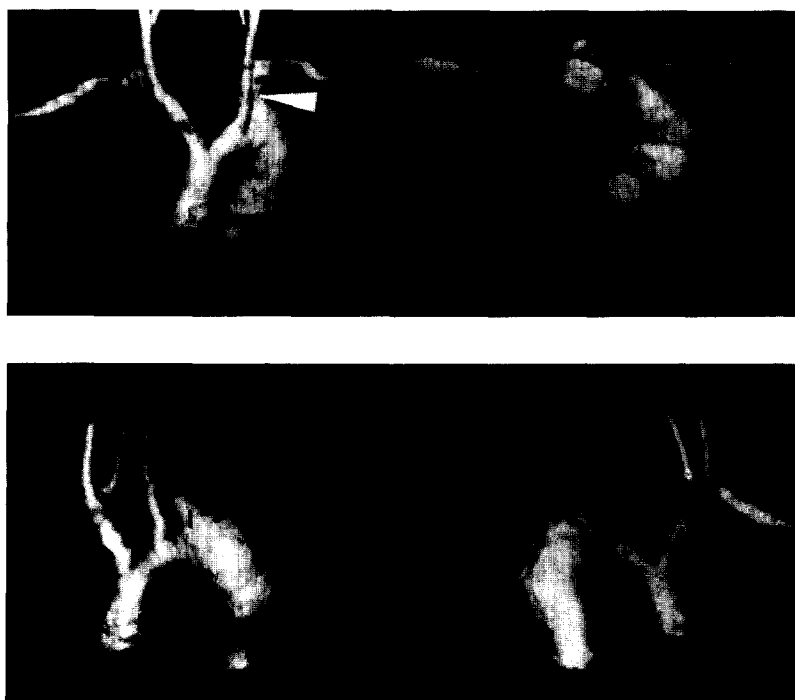
rupture. Rarely were complaints of chest pain or discomfort reported. Patients without rupture, despite the presence of extensive aneurysmal dilation, were usually asymptomatic or had rare complaints of paraspinal discomfort. Chest x-ray findings were often abnormal, with an enlarged aortic knob, which was noted in patients with a large aneurysm as well as in those with mild to marginal dilation.

Pregnancy. Four of the five pregnant patients developed aneurysms, and rupture occurred in three of these four. The majority of aneurysms occurred in female patients, which leads us to consider that female gender carries a higher risk despite the absence of statistical significance in the present study. Pregnancy appears notably to increase the risk of aneurysm rupture. Although the underlying mechanism for this finding is not clearly understood, it might be attributed to increased cardiac output and volume and pressure overload during pregnancy (12-15).

Pregnancy as a risk factor for patients of child-bearing age should prompt early evaluation before a planned conception and shortly after delivery. Studies during the course of pregnancy by other noninvasive methods or techniques might become necessary if deemed clinically warranted. Several patients with ruptured aneurysms complained previously of hemoptysis and paraspinal back pain as their only reported symptom (16), and despite an apparent higher incidence in women, no statistical difference between the male and female patients was found in the incidence of aneurysm formation as related to age at surgical repair.

Hypertension. The number of hypertensive patients in the present study (11 [39%] of 39) was consistent with that previously documented in population studies after patch repair (17). It occurred with similar frequency in our patients, both

Figure 3. Three-dimensional surface image of a 21-year old man 15 years after Dacron patch aortoplasty, with a large saccular aneurysm (arrow) shown in multiple views. This patient has suspected aneurysmal involvement of the base of the left subclavian artery (I) as well; the extension of the repair onto the base of the subclavian artery occurred over several repairs to place larger patches. The other brachiocephalic vessels are free of involvement. The descending aorta also illustrates the most inferior extension of the lesion.



those with aneurysms and those free of aneurysmal dilation after Dacron patch aortoplasty. Hypertension did not appear to be a major factor in the present study, but its etiology should be determined and therapy instituted because its possible contribution to aneurysm formation and rupture cannot be dismissed (3,18).

Pathologic findings. Medial fibrosis with disruption of the elastic lamellae and peripatch atherosclerotic changes appeared in several patients studied after aortic rupture or aneurysm resection. Intimal thickening and fibrosis with absence of an identifiable medial layer was also reported in one patient, again with atherosclerotic changes. The common factor identified among the patients with aneurysm formation was disruption of the integrity of the aortic layers in the peripatch area. This disruption appears to contribute to weakness of the vessel at this point. This segment of the aorta may also have an abnormal wall structure by virtue of the original coarctation and adjacent ductal tissue.

Discussion

In 1983, Ala-Kulju et al. (4) reported a 27% incidence of aneurysm formation after patch aortoplasty in adult patients. In 1988, the same group reported a 32% incidence and suggested that the incidence (5) and rate (19) of aneurysm formation increased over time with continued follow-up. Our findings, which reveal an overall 51% incidence of aneurysm after patch aortoplasty with almost 17 years of follow-up, strongly support those observations.

Aneurysm incidence in group I is much lower than that in group II (11% vs. 63%), even though survival rates for both groups were not significantly different (88.89% vs. 83.33%). The high survival rate for group II was partially attributed to timely detection of aneurysms and subsequent surgical resection. Mortality rate conceivably would have increased had detection and intervention not been prompted by the present study. A low incidence of postpatch aneurysm formation for this procedure in group I allowed us to speculate that Dacron patch aortoplasty repair of coarctation of the aorta in children <2 years old may prove in long-term follow-up to be a reliable surgical approach. This operation has an unfavorable outcome when performed in patients >2 years of age. Follow-up at regular intervals is recommended in either case.

Aneurysm formation (suspected etiologies). Causative factors of aneurysm development appear to include the utilization of larger patches (20) and associated increased wall stress in the repair of the older group, and should include the higher relative blood pressure in older patients than in newborns and infants. Other factors to be considered are the higher elasticity of the aorta in newborns and infants and surgical removal of the anterior intimal ridge.

Age risk factor. The present study clearly demonstrates a specific time frame, <2 years of age, at which Dacron patch aortoplasty should be performed. Previous studies suggested that this technique be performed as early as possible (21,22) and abandoned in adolescents and adults (10) but never



Figure 4. Static spin-echo sagittal image showing a large saccular aneurysm. Hemorrhage (h) shown over apical portion of aneurysm with some posterior and lateral extension. The ascending (a) and descending aortas (d) are normal, with no aneurysmal involvement. A = anterior; P = posterior.

specifically suggested a cutoff time frame. Thus, the continued use of this surgical technique may have a role in younger patients at those institutions that continue to practice patch aortoplasty as a primary surgical approach to correction of coarctation of the aorta (23).

Noninvasive imaging. Magnetic resonance imaging has gained popularity for its safety and accuracy in aneurysm detection in both adults (24) and children (25) and has proved to be an important noninvasive and economically competitive method for evaluation of patients with suspected or known aortic pathology. Our experience suggests that a complete MRI examination, including static spin-echo images and cine fast gradient echo images, should be performed in all patients with previous Dacron patch aortoplasty repair of coarctation of the aorta. Additional computer-generated three-dimensional reconstructed images are very helpful for presurgical planning and can be developed from both the contiguous multislice gated inflow or transverse spin-echo images. Surgeons at Emory resected aneurysms in nine patients relying solely on information provided by these three-dimensional reconstructed angiographic images and found these images to be extremely accurate and precise renderings of the anatomy found at operation.

Conclusions. Late aneurysm formation may be a potentially lethal complication of Dacron patch aortoplasty repair of coarctation of the aorta. It is more likely to occur in female patients operated on >2 years of age and especially among women who become pregnant. The risk of aneurysm formation appears to be substantially reduced by operating earlier (<2 years of age), yielding 63% versus 11% in this study. Female patients may have a higher incidence of aneurysm formation

after Dacron patch aortoplasty than their male counterparts (70% vs. 39% in this study, but $p > 0.05$), and pregnancy appears to be a significant late risk factor for subsequent rupture.

Magnetic resonance imaging with magnetic resonance angiography and three-dimensional surface rendering provides a noninvasive, radiation-free method of obtaining high resolution images of the entire thoracic aorta and adjacent structures. In addition, three-dimensional MRI (reconstructed images) allows display of the aorta in any desired perspective and has proved to be valuable to surgeons in planning the course of operation and in reviewing postoperative results. Static spin-echo MRI also provides multiplanar review and, in cases where rupture is suspected, may delineate hemorrhage (Fig. 4). In addition, detailed segmental soft tissue anatomy is clearly provided. Because of these features and demonstrated accuracy in three-dimensional anatomic depictions, static MRI studies and magnetic resonance angiography with three-dimensional reconstruction are indeed preferred over other imaging methods by surgeons in our institution as a definitive evaluation procedure before resection of aortic aneurysms.

References

- Vosschulte K. Isthmusplastiek zur Behandlung der Aortem Isthmusstenose. *Thoraxchirurgie* 1957;4:443-50.
- Pennington DG, Liberthson RR, Jacobs M, Scully H, Goldblatt A, Daggett VM. Critical review of experience with surgical repair of coarctation of the aorta. *J Thorac Cardiovasc Surg* 1979;77:217-29.
- Bergdahl L, Ljungquist A. Long term results after repair of coarctation of the aorta by patch grafting. *J Thorac Cardiovasc Surg* 1980;80:177-81.
- Ala-Kulju K, Jarvinen A, Maamies T, Mattila S, Merikallio E. Late aneurysm after patch aortoplasty for coarctation of the aorta in adults. *Thorac Cardiovasc Surg* 1983;31:301-5.
- Ala-Kulju K, Heikkinen L. Aneurysm after patch graft aortoplasty for coarctation of the aorta: long term results of surgical management. *Ann Thorac Surg* 1989;47:853-6.
- Messmer BJ, Minale C, Muhler E, Von Bernuth G. Surgical correction of coarctation of the aorta in early infancy: does surgical technique influence the result? *Ann Thorac Surg* 1991;52:594-603.
- Pinzon JL, Burrows PE, Benson LN, et al. Repair of coarctation of the aorta: post-operative morphology. *Radiology* 1991;180:199-203.
- Clarkson PM, Brandt PWT, Barrat-Boyes BG, Rutherford JD, Kerr RA, Neutze JM. Prosthetic repair of coarctation of the aorta with particular reference to Dacron onlay patch grafts and late aneurysm formation. *Am J Cardiol* 1985;56:342-6.
- Bromberg BI, Beckman RH, Rocchini AP, et al. Aortic aneurysms after patch aortoplasty repair of coarctation of the aorta: prospective analysis and prevalence. *J Am Coll Cardiol* 1989;14:734-41.
- Albert H, Bednarski P, Laas J, Koch U, Prokol M, Borst HG. High incidence of aneurysm formation following patch plasty repair of coarctation. *Eur J Cardiothorac Surg* 1993;7:200-5.
- de Graff RG, Groen JP. Magnetic resonance angiography with pulsatile flow. *Mag Res Imag* 1992;10:25-34.
- Chesley LC. Plasma and red cell volume during pregnancy. *Am J Obstet Gynecol* 1972;112:440-50.
- Metcalf J, Ueland K. Maternal cardiovascular adjustment to pregnancy. *Prog Cardiovasc Dis* 1974;16:363-74.
- Lindeheimer MD, Katz AI. Sodium and diuretics in pregnancy. *N Engl J Med* 1973;288:891-4.
- Rubler S, Damani PM, Pinto ER. Cardiac size and performance during pregnancy: estimation with echocardiography. *Am J Cardiol* 1977;40:534-40.
- Holdright DR, Kilner P, Somerville J. Hemoptysis from false aneurysm: near fatal complication of repair of coarctation of the aorta using a Dacron patch. *Int J Cardiol* 1991;32:406-8.
- Malan JE, Benata A, Levin SE. Long term follow-up of coarctation of the aorta repaired by patch angioplasty. *Int J Cardiol* 1991;30:23-32.
- Olsson P, Soderlund S, Dubiel WT, Ovenfors CO. Patch graft or tubular grafts in the repair of coarctation of the aorta: a follow-up story. *Scan J Thorac Cardiovasc Surg* 1976;10:139-43.
- Mendelson A, Crowley D, Lindener A, Beekman R. Rapid progression of aortic aneurysm after patch aortoplasty repair of coarctation. *J Am Coll Cardiol* 1992;20:381-5.
- Del Nido PJ, Williams WG, Wilson GJ, et al. Synthetic patch angioplasty for repair of coarctation of the aorta: experience with aneurysm formation. *Circulation* 1986;74(Pt 2):132-6.
- Cohen M, Fuster V, Steele PM, Driscoll D, McGoon DC. Coarctation of the aorta: long term follow-up and prediction of outcome after surgical correction. *Circulation* 1988;80:840-5.
- Presbitero P, Demarie D, Villani M, et al. Long term results (15-30 years) of surgical repair of aortic coarctation. *Br Heart J* 1987;57:462-7.
- Bertolini A, Dalmonte P, Toma P, et al. Goretex patch aortoplasty for coarctation in children: nuclear magnetic resonance assessment at 7 years. *Cardiovasc Surg* 1992;33:223-8.
- Rofsky NM, Weinreb JC, Grossi EA, et al. Aortic aneurysm and dissection: normal MR imaging and CT findings after surgical repair with continuous suture graft inclusion technique. *Radiology* 1993;186:195-201.
- Bank ER, Aisen AM, Rocchini AP, Hernandez RJ. Coarctation of the aorta in children undergoing angioplasty: pretreatment and posttreatment magnetic resonance imaging. *Radiology* 1987;162:235-40.